

Three unusual parapharyngeal space masses resected via the endoscopy-assisted transoral approach: case series and literature review

Journal of International Medical Research

48(6) 1–6

© The Author(s) 2020

Article reuse guidelines:

sagepub.com/journals-permissions

DOI: 10.1177/0300060520936068

journals.sagepub.com/home/imr

Li-Fang Shen , Ya-Lian Chen and Shui-Hong Zhou 

Abstract

Tumors of the parapharyngeal space (PPS) are rare, most originate from salivary and neurogenic tissues, and most are benign. However, there are some rarer masses in the PPS, with just a few published reports in the literature worldwide, and we may not consider them in the differential diagnosis of PPS neoplasms. We report three cases of rare masses in the PPS: Warthin's tumor, branchial cleft cyst, and carcinoma ex pleomorphic adenoma. The three patients were admitted to our department with complaints of painless swelling in the lower side of the right face or a long history of snoring; diagnoses were confirmed histopathologically. An endoscopy-assisted transoral approach was used that allowed wide visibility for safe resection and resulted in a short hospitalization time and good functional and cosmetic outcomes. All patients have been followed to the current time, and there have been no recurrences. The transoral endoscopy-assisted approach appears to be safe, effective, and less invasive for excision of masses in the PPS.

Keywords

Parapharyngeal space tumors, Warthin's tumor, branchial cleft cyst, carcinoma ex pleomorphic adenoma, endoscopy-assisted operation, transoral approach

Date received: 3 February 2020; accepted: 29 May 2020

Department of Otolaryngology, The First Affiliated Hospital, College of Medicine, Zhejiang University, Hangzhou City, China

Corresponding author:

Li-Fang Shen, Department of Otolaryngology, The First Affiliated Hospital, College of Medicine, Zhejiang University, 79 Qingchun Road, Hangzhou City, Zhejiang 310003, China.

Email: 11418276@zju.edu.cn



Creative Commons Non Commercial CC BY-NC: This article is distributed under the terms of the Creative Commons Attribution-NonCommercial 4.0 License (<https://creativecommons.org/licenses/by-nc/4.0/>) which permits non-commercial use, reproduction and distribution of the work without further permission provided the original work is attributed as specified on the SAGE and Open Access pages (<https://us.sagepub.com/en-us/nam/open-access-at-sage>).

Introduction

Tumors of the parapharyngeal space (PPS) are rare, accounting for 0.5% of head and neck carcinomas. Moreover, 70% to 80% of PPS tumors are benign and most originate from salivary and neurogenic tissues.¹ In addition to these common tumors, some rare tumors occur in the PPS. Warthin's tumor (WT), carcinoma ex pleomorphic adenoma (CXPA), and second branchial cleft cyst are rarely encountered in the PPS. The complexity of anatomy, varied treatment approaches, and atypical presentations make these tumors worthy of study and report.

Case reports

We retrospectively collected three unusual PPS masses: WT, branchial cleft cyst, and CXPA, which were completely resected via an endoscopy-assisted transoral approach (EATA) under general anesthesia by Dr. Zhou and his medical team. All procedures were in accordance with the ethical standards of the responsible committee on human experimentation (the First Affiliated Hospital, College of Medicine, Zhejiang University, China, reference number 2018-1019) and with the Declaration of Helsinki of 1975, as revised in 2008. Informed consent was obtained from all patients included in the study.

Case 1

A 72-year-old male patient presented with a painless swelling on the right side of his face that had been present for 1 year in January 2018. A submucosal soft mass was observed under the right lateral wall of the oropharynx. The patient had been a heavy smoker for the last 30 years and had no relevant family history. Contrast-enhanced magnetic resonance imaging (MRI) revealed a 34 × 29 × 23-mm well-defined mass arising from the deep lobe of parotid gland, with

T1 hypointense contents and T2 hyperintense contents with mixed signal inside; postcontrast enhancement was significant (Figure 1a, b). The final pathological result confirmed the diagnosis of Warthin's tumor.

Case 2

A 32-year-old male presented with a 3-year history of snoring and a feeling of numbness in the right laterocervical region that had been present for 6 months in November 2014. A soft painless mass under the right lateral pharyngeal wall was observed. MRI revealed a 54 × 34 × 23-mm cystic mass in the right PPS (Figure 1c, d), which was compressing the right tonsil anteriorly, causing severe narrowing of the oropharyngeal and nasopharyngeal passage. The histopathological diagnosis was a branchial cleft cyst.

Case 3

A 61-year-old man presented with a 6-month history of snoring in September 2015. There was no facial paralysis, nasal congestion, sore throat, or dysphagia. A mass under the left nasopharyngeal wall was observed by endoscopy. MRI revealed a 50 × 27 × 49-mm irregular mass in the right PPS, with T1 equisignal contents, T2 hyperintense contents, and asymmetrical postcontrast enhancement (Figure 1e, f), without indication of lymph node enlargement. The postoperative pathological analysis revealed CXPA and the resection margins were free of carcinoma. After surgery, the patient received local radiotherapy, DT 5000 cGy/25F. The patient underwent MRI regularly during follow-up and there was no tumor recurrence or complications.

Total excision of the masses in all three patients was performed via an EATA with or without splitting of the soft or hard palate, according to the size and location

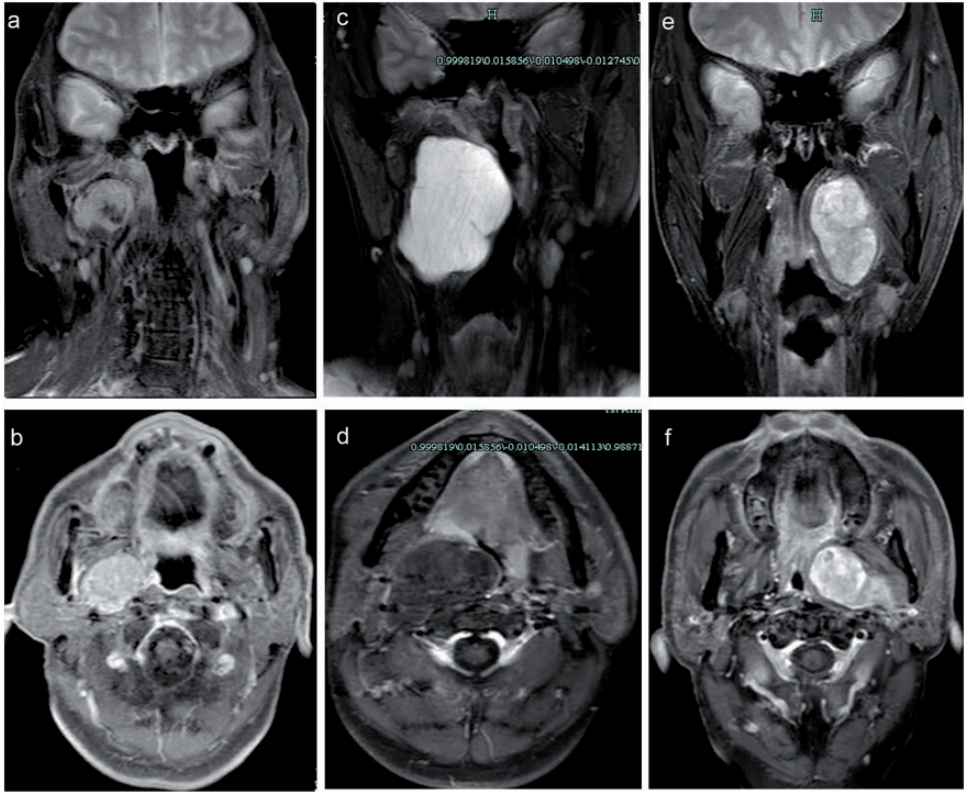


Figure 1. Contrast-enhanced magnetic resonance imaging (MRI) of the three cases. (a) Case 1: MRI revealed a $34 \times 29 \times 23$ -mm well-defined mass arising from the deep lobe of parotid gland, showing T2 hyperintense contents with mixed signal inside; (b) postcontrast enhancement was significant. (c) Case 2: MRI revealed a $54 \times 34 \times 23$ -mm cystic mass in the right parapharyngeal space with T2 hyperintense contents; (d) no significant postcontrast enhancement was seen. (e) Case 3: MRI revealed a $50 \times 27 \times 49$ -mm irregular mass in the right parapharyngeal space with T2 hyperintense contents; (f) asymmetrical postcontrast enhancement was seen.

of the tumor (Karl Storz Endoskope system with a 0 degree endoscope, 4-mm diameter, 30 cm; Karl Storz SE & Co. KG, Tuttlingen, Germany). An incision was made with an electric scalpel on the palatopharyngeal arch, and the tumor was removed using blunt resection under endoscopy. Endoscopy made the operation field clear and effectively protected important surrounding blood vessels and nerves during the operation. No neurovascular injury occurred in the process, and endoscopy helped us avoid complications

(Figure 2). Table 1 summarizes the clinical and surgical characteristics of the three cases.

Discussion

Tumors that originate in the PPS are rare and their histopathology is varied; about 70 histopathologic types in PPS are reported.² WT was first described by Hildebrand in 1895; Warthin described similar cases and the tumor was named after him in 1929. WT almost always originates in the parotid

gland, and extra-parotid presentations are very rare.³ The main histologic characteristic of this tumor is its abundant lymphoid stroma, the origin of which is the subject of debate. It is unclear whether the lymphoid tissue is a reactive or neoplastic response. Patients with WT are more likely to be heavy smokers than patients with other salivary tumors.⁴ The probability of transformation of WT into carcinoma or malignant lymphoma is 0.3% to 1.1%;⁵ thus, rapid growth of a long-standing WT may indicate a malignant transformation.

The second branchial cleft cyst may arise anywhere from the tonsillar fossa to the supraclavicular area. It is extremely

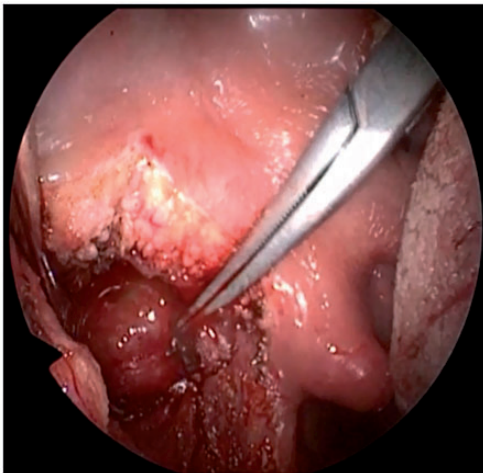


Figure 2. Excision of the mass via an endoscopy-assisted transoral approach.

uncommon in PPS. Diverse approaches have been implemented to treat parapharyngeal cysts, including drainage and incision, injection of sclerosant substances, and marsupialization. Nevertheless, these methods do not always guarantee long-term efficacy, and surgical excision remains the only definitive solution.

CXPA is an uncommon malignancy of the salivary gland, and it comprises approximately 5% to 15% of all salivary tumors.⁶ CXPA can be composed of diverse histologic subtypes, including adenocarcinoma not otherwise specified, high-grade salivary duct carcinoma, sarcomatoid carcinoma, and myoepithelial carcinoma. Precise preoperative diagnosis of CXPA is almost impossible. The 5-year survival rate of CXPA ranges from 25% to 75%, and CXPA is considered to have one of the worst prognoses of all parotid tumors.⁶

The major goals of PPS tumor surgery are to completely remove the tumor and reduce the main postoperative complications. Classic methods of approach for PPS tumors include transparotid, transcervical, transmandibular, and combinations thereof. The choice of approach is mainly based on the site, vascularity, size, and histology of the tumor, especially the relationship between the carotid sheath and the tumor. It should provide wide visibility for safe resection during the operation with minimal functional or cosmetic sequela. In our cases, the transoral approach provided

Table 1. Clinical and surgical information data from the three cases.

Case No	Sex	Age (years)	Surgery time (minutes)	Tumor size (mm)	Pathologic diagnosis	Bleeding (mL)	Hospitalization time (days)	Recurrence or complication	Follow-up time (months)
1	Male	72	60	35 × 30 × 20	Warthin's tumor	50	5	No	23
2	Male	32	80	54 × 34 × 23	Branchial cleft cyst	50	5	No	61
3	Male	61	89	60 × 45 × 30	CXPA	50	7	No	51

CXPA, carcinoma ex pleomorphic adenoma.

a safe and wide visible approach to the PPS; this results in a lower rate of postoperative complications and has some advantages.⁷ Betka et al.¹ recommended this approach because of its superior visual control of dissection over the upper and middle parts of the tumor, good cosmetic and functional outcomes, and short surgical and hospitalization times. The main limitation of the transoral approach is the narrowed access, which may hamper complete tumor resection and increase the risk of neurovascular injury; however, these limitations can be overcome by the endoscopy-assisted approach.

The EATA can provide a sufficient surgical window for tumor removal from the PPS, and the important anatomical structures near the tumor, including the carotid sheath and IX-XII cranial nerves, can be clearly exposed and protected via the endoscopy-assisted approach. This approach reduces the risk of inferior alveolar nerve and facial nerve injury and salivary fistulae, eliminates parotid dissection and obvious facial scars, and achieves excellent functional and aesthetic results.⁸ Wang et al.⁹ found that there was no significant difference in complication rate, operation time, total removal rate, or recurrence rate between EATA and external approaches. However, EATA has advantages of less postoperative pain level, reduced blood loss, preservation of facial appearance and function, and shortened hospitalized time.⁹ However, because of the rigid bars of the harmonic scalpel, some inferior anatomic fields cannot be reached with harmonic scalpels during EATA. Therefore, external approaches or robotic technologies should be considered in these cases. In our study, all of the tumors were removed completely by EATA, and blood vessels and nerves were carefully preserved during the separation process.

In conclusion, WT, CXPA, and second branchial cleft cyst in the PPS are

rarely encountered. Although surgical approach remains a challenge, EATA provides better functional and aesthetic results than external approaches in selected cases.

Acknowledgements

We acknowledge the assistance of all those who participated in this project, especially the technologists in the Pathology department of the First Affiliated Hospital, College of Medicine, Zhejiang University.

Declaration of conflicting interest


The authors declare that they have no conflict of interest.

Funding

This research was supported by Science and Technology Department of Zhejiang Province, China (No. 2016C33144).

ORCID iDs

Li-Fang Shen  <https://orcid.org/0000-0002-2245-8544>

Shui-Hong Zhou  <https://orcid.org/0000-0002-7163-2289>

References

1. Betka J, Chovanec M, Klozar J, et al. Transoral and combined transoral-transcervical approach in the surgery of parapharyngeal tumors. *Eur Arch Otorhinolaryngol* 2010; 267: 765–772.
2. Riffat F, Dwivedi RC, Palme C, et al. A systematic review of 1143 parapharyngeal space tumors reported over 20 years. *Oral Oncol* 2014; 50: 421–430.
3. Shaw CK, Sood S, Bradley PJ, et al. Unusual mass in the parapharyngeal space: a Warthin's tumour. *ANZ J Surg* 2006; 76: 193–194.
4. Ethunandan M, Pratt CA, Higgins B, et al. Factors influencing the occurrence of multicentric and 'recurrent' Warthin's tumour: a cross sectional study. *Int J Oral Maxillofac Surg* 2008; 37: 831–834.
5. Gunduz M, Yamanaka N, Hotomi M, et al. Squamous cell carcinoma arising in a

- Warthin's tumor. *Auris Nasus Larynx* 1999; 26: 355–360.
6. Zbären P, Zbären S, Caversaccio MD, et al. Carcinoma ex pleomorphic adenoma: diagnostic difficulty and outcome. *Otolaryngol Head Neck Surg* 2008; 138: 601–605.
 7. Fan S, Lin SG, Zhang HQ, et al. A comparative study of the endoscopy-assisted transoral approach versus external approaches for the resection of large benign parapharyngeal space tumors. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2017; 123: 157–162.
 8. Chen WL, Fan S, Huang ZQ, et al. Endoscopy-assisted transoral versus endoscopy-assisted transcervical minimal incision plus mandibular osteotomy approach in resection of large parapharyngeal space tumors. *J Craniofac Surg* 2017; 28: 976–979.
 9. Wang X, Gong S, Lu Y, et al. Endoscopy-assisted transoral resection of parapharyngeal space tumors: a retrospective analysis. *Cell Biochem Biophys* 2015; 71: 1157–1163.